

Holmium laser endourethrotomy and litholapaxy of an occult prostatic utricle calculus

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Prostatic utricle (PU) stones are rare and the surgical treatment of such PU anomalies has proved challenging given their location adjacent to the posterior urethra and proximity to important fertility and continence structures. We report the case of a 58-year-old male with hypospadias,

microphallus, and irritative lower urinary tract symptoms (LUTS) found to have a large PU stone. He underwent successful complete endoscopic removal of the stone via transurethral unroofing and holmium laser litholapaxy. We conclude that it is an effective alternative to other surgical techniques once size of the stone and the patient's individual anatomy are considered.

Key Words: urethrolitholapaxy, endoscopy, prostate, prostatic stone

Introduction

It has been estimated that about 51% of middle-aged men have a prostatic stone.¹ Stones of the prostatic utricle (PU), however, are rare. The first reported case of two children with PU stones was published in 1958 and since that time only three other reports involving 12 individuals with PU stones have been reported.^{2,3} Presence of a PU in the general population occurs in 4% of newborns and 1% of adults, while presence of a PU in individuals with hypospadias or intersex disorders is increased at 11%-14%.⁴ The etiology of PU stones is unclear, but as with other prostatic calculi, they are thought to represent a dystrophic, benign,

inflammatory-mediated process due to precipitation of prostatic secretions and calcification of the corpora amylacea.⁵ As with other prostatic stones, PU stones can be asymptomatic or associated with irritative lower urinary tract symptoms (LUTS), infection, and urinary retention depending on size and degree of mechanical obstruction or inflammation by the stone. Retrograde urethrography (RUG) is the most sensitive method for detecting an enlarged PU.⁶ Symptomatic PU abnormalities require surgical intervention for definitive management. However, the surgical management of PU stones has proven difficult due to the rarity of the problem, difficulty of obtaining access to the posterior urethra, and the close anatomic relationships to important structures required for fertility and continence. Open, laparoscopic, and endoscopic approaches have been described for excision and removal of Müllerian duct abnormalities; however literature on the surgical removal of PU stones is sparse.

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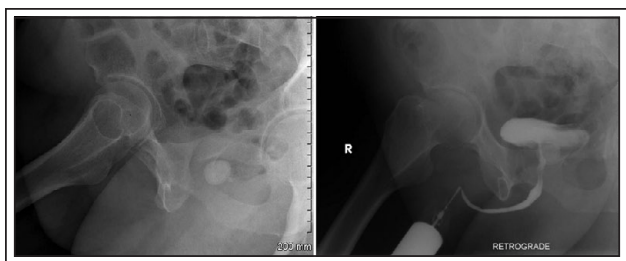


Figure 1. Retrograde urethrogram with scout film demonstrating utricular stone immediately posterior to normal urethra.

Case report

We report the case of a 58-year-old man with a PU stone, hypospadias, and irritative LUTS managed with transurethral holmium laser urethrolitholapaxy for a stone filling a dilated Müllerian remnant causing LUTS, dysuria, and recurrent urinary infection. Past surgical history included bilateral inguinal hernia repair and left testicular repair for possible torsion. Physical examination revealed a microphallus with subcoronal hypospadias and a nontender 2 cm mass contiguous with the posterior edge of his diminutive prostate. Labs were consistent with hypogonadism, revealing a low testosterone of 62 ng/dL, elevated FSH of 31.7 mIU/mL, normal LH of 8.9 mIU/mL, and normal prolactin of 7 ng/mL. PSA was low at < 0.1 ng/mL and Cr was 1.06 mg/dL. Uroflowmetry and post-void residual (PVR) demonstrated a peak flow of 6 mL/sec and a PVR of 7 mL, respectively. Retrograde urethrogram identified a calcification immediately posterior to his otherwise normal prostatic urethra, Figure 1. Computed



Figure 2. Computed tomography demonstrated a large midline calcification posterior to the prostatic base.

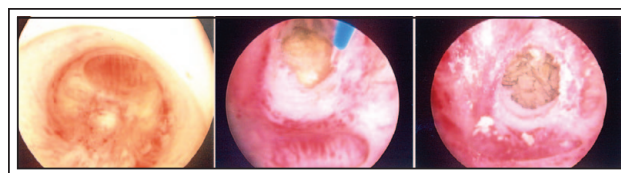


Figure 3. a) Pinpoint utricular ostium with a midline submucosal calculus; b) Holmium laser unroofing of prostatic utricle; c) In situ laser lithotripsy of utricle stone.

tomography demonstrated a large midline calcification posterior to the prostatic base, Figure 2. Office cystoscopy was not possible due to meatal stenosis.

Cystoscopy under anesthesia using a 12F rigid cystoscope revealed a pinpoint utricular ostium with a midline submucosal calculus, Figure 3a. The bladder was inspected and found to be without lesions, masses, or stones. Inspection of the prostatic urethra did not show an obstructing prostate or evidence of benign prostatic hyperplasia (BPH). A 365 micron holmium laser fiber with laser settings of 0.8 joules and 8 Hertz was used to incise the prostatic urethra, thus marsupializing the dilated utricle into the prostatic urethra, Figure 3b. The 2 cm stone was identified, fragmented in situ, Figure 3c, and extracted with rigid graspers. A 16 French Foley catheter was left in place for 2 weeks. Stone analysis revealed a weight of 1.082 g and a composition of 60% calcium phosphate (apatite) and 40% ammonium urate. Postoperatively, the patient has been voiding without recurrent symptoms or urinary tract infections.

Discussion

Utricular anomalies can occur from incomplete regression of the Müllerian ducts or incomplete closure of the urogenital sinus. In normal embryogenesis, the Müllerian ducts regress due to the influence of anti-Müllerian hormone (AMH) secreted by normally functioning fetal testes. Utricular anomalies are found more commonly in individuals with hypospadias or intersex disorders and a direct relationship has been shown between the severity of hypospadias and the degree of utricular enlargement.^{2,7}

Although the true etiology of PU stones is unclear, they are thought to occur via the same etiology as other prostatic calculi. A recent study that reviewed the composition of PU stones determined them to be made primarily of hydroxyapatite crystal, possibly from a mix of concentrated liquid secreted by the PU and deciduous epithelial cells.² Individuals with PU stones may present with persistent LUTS, urinary

retention, or recurrent UTIs. Surgical intervention is the definitive treatment for symptomatic PU anomalies including enlarged PUs, PU cysts, and PU stones. While there is extensive literature on surgical management of enlarged PUs and Müllerian duct cysts in children, management of PU stones in adults has been described rarely.

Open transvesical transtrigonal, suprapubic, extravesical, transrectal, and more recently, laparoscopic and endoscopic techniques have all been described for surgical management of Müllerian duct remnants.⁸⁻¹⁰ With any technique, complete exposure, visualization, and removal of the PU without damage to the urethra, external sphincter, or branches of the pelvic plexus is the goal. The transvesical transtrigonal approach has been reported to enable visualization of both the utricle and the vas deferens with simple technique and minimal complications. The suprapubic extravesical approach allows easy exploration of other pelvic organs and avoids opening of the bladder. However, it involves extensive dissection, difficulty in mobilizing the rectum, and limited exposure due to a narrow working field. The perineal approach is considered to be technically difficult and puts the external sphincter and rectum at greater risk.

While an open approach is recommended by some to be superior in the pediatric population, advantages of endoscopy include direct visualization of the posterior urethra, avoidance of rectum and bladder excision, minimal postoperative morbidity, good wound cosmesis, and decreased length of hospital stay. However, endoscopic instrumentation can be limited by urethral anatomy and it is recommended that individuals with residual female remnants including a true vagina, fallopian tubes, or uterus undergo open repair. Limitations of endoscopic electrofulguration include the possibilities of persistent utricular remnants leading to recurrent symptoms and electrical current damage to the ejaculatory ducts leading to infertility.

In choosing the best surgical approach for our patient, we took into account his small urethral meatus and microphallus, the location of the ostium at the verumontanum, and size of his stone. Given these findings, we found an endoscopic transurethral approach with laser incision of the posterior urethra and urethrolitholapaxy via holmium laser to be the best management. Although electrofulguration for enlarged PUs or PU cysts has been described and open or endoscopic removal of PU stones is reported, we report the first case of endoscopic urethrolitholapaxy for removal of a PU stone. Consideration of the size of the stone and individual anatomy must be addressed in choosing a surgical approach for removal. □

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